Crico Arytenoid Joint Fixation in Diffuse Idiopathic Skeletal Hyperostosis (DISH): A Case Report

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Abstract Advanced form of diffuse idiopathic skeletal hyperostosis or Forestier's disease can induce dysphagia and significant airway symptoms such as hoarseness, snoring, dyspnoea on exertion and laryngeal stridor. We have discussed the diagnosis and management of an unusual case with respiratory distress due to left cricoarytenoid joint fixation and right vocal cord paresis in conjunction with skeletal pathology.

Keywords Diffuse idiopathic skeletal hyperostosis (DISH) · Forestier's disease · Vocal cord immobilisation

Introduction

Forestier's disease or diffuse idiopathic skeletal hyperostosis is an ossifying diathesis, characterized by paravertebral osteophyte formation, calcification and ossification of the anterior longitudinal vertebral ligament [1]. It is a rheumatic abnormality leading to exuberant proliferation of bone at different osseous sites of ligaments and tendinous attachment throughout the body. The disease is most common in middle age and the elderly. It has also been described in younger age groups with a predilection in

males [2]. About 28% of patients with DISH related cervical osteophytosis present with dysphagia [3]. On the contrary laryngo tracheal symptoms like dyspnoea, snoring, stridor and vocal cord adduction immobilization are rare [4]. A case of progressive chronic dyspnoea on exertion due to unilateral cricoarytenoid joint fixation and bilateral edematous arytenoids associated with DISH which has not been published in the literature is being reported.

Case Report

A 65 year old male visited our institution with severe stridor on exertion and dysphagia for 3 years. He complained of dry cough, change of voice for 1 year. He had undergone cervical osteophyte excision by left anterior cervical approach for relief of dysphagia before 4 years. His past medical records revealed that vocal cord mobility was normal following the procedure. Flexible nasopharyngoscopy revealed an immobile left vocal cord in a median position with restricted right vocal cord movements. The osteophytic mass started at the level of oropharynx in the posterior pharyngeal wall and stopped short of the arytenoids with mucosal odema of both arytenoids. Glottic chink was found to be 4 mm on maximum inspiration. Under local anaesthesia a blunt curved laryngeal biopsy forceps was passed via the mouth and left arytenoid palpated and found to be fixed. Right arytenoid was found to be mobile. Plain radiographic examination (Fig. 1) of the cervical spine revealed significant bony osteophytic bridges extending anteriorly between C4 and C7. CT of the neck revealed large anterior osteophytes, involving C4 to C7 vertebral bodies causing compression of the posterior aspect of larynx. Xray of the dorso lumbar spine confirmed the radiologic findings of DISH.

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Fig. 1 Pre op X-ray cervical spine–skeletal hyperostosis

Surgical Treatment

Trans Cervical Excision of Cervical Osteophytes with Left Arytenoidectomy and Cordopexy

After a preliminary tracheostomy, a right sided horizontal skin crease incision at the level of thyroid cartilage was placed, the soft tissues were retracted after identifying the omohyoid deeper to which the vertebrae were reached.

The anterior longitudinal ligament was found to be calcified with gross anterior beaking at the level of C3 and C5. Both areas were drilled and smoothened under flexible laryngoscopic control. The incision was extended on to the left side, strap muscles cut, to identify the posterior border of thyroid cartilage and a Maragos window created. The ossified arytenoid cartilage with the fixed, immobile cricoarytenoid joint was identified by following the PCA muscle. The joint space was separated, and the arytenoid

excised after performing a submucosal dissection, along with the corniculate cartilage. Lateralization of the vocal process by a prolene stitch anchored laterally and inferiorly to the inferior cornu of thyroid cartilage was executed and the wound closed in layers [5]. The tracheostomy was allowed to close after 5 days.

On follow up (Fig. 2) 2 months later the glottic airway was greater than 8–10 mm on inspiration, no stridor during inspiration and the left vocal cord was lateralised. Right vocal cord was found to be mobile. In addition minor prolapse of soft tissue from the area of left arytenoid into the glottic inlet on inspiration was noted (Fig. 2a).

The mucosal prolapse increased and led to stridor and 5 months later the patient underwent microdebrider excision of the prolapsing mucosa. He was asymptomatic, and again 1 year later acquired inspiratory stridor, this time due to supraglottic mucosal prolapse. LASER vapourisation of the supraglottic mucosa was done and he has since been asymptomatic on follow up for the last 2 years with an adequate glottis (Fig. 2b Final Airway).

Discussion

This patient had left cricoarytenoid joint fixation and right sided vocal paresis. The ankylosis of cricoarytenoid joint may be due to mechanical compression by a prevertebral hyperostosis, which subsequently causes chondritis of the cricoid cartilage and arytenoid with ankylosis of the cricoarytenoid joint [6–8]. This patient also had edematous swelling of the laryngeal structures and right vocal paresis. Compression of the venous structures in this area occurs by hyperostosis at the thoracocervical level, consequently this may also be responsible for inducing edema, fibrosis and immobilization of the arytenoids [9]. because of the

Fig. 2 a Post operative after 2 months b: Image seen after LASER vapourisation of prolapsing supraglottic mucosa

cricoarytenoid joint fixation this patient underwent arytenoidectomy along with vocal cord lateralization in addition to the cervical decompression. The right vocal paresis improved spontaneously following treatment. This was probably consequent to the improvement in laryngeal edema following osteophyte removal and also proves the normalcy of the right recurrent laryngeal nerve. Laryngeal EMG could not be done as the thyroid cartilage was ossified and the cricothyroid membrane was scarred because of previous surgery [10].

Conclusion

Respiratory distress in DISH was thought to be due to venous compression, nerve compression, cricoarytenoid joint fixation, post cricoid ulceration and odema. We have reported the first documented case with cricoarytenoid joint fixation as a cause of stridor and methods to identify the problem in clinical setting, when EMG is not feasible. Treatment of the coexistent laryngeal pathology is necessary in such a presentation.

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